

Dupuytren's Contracture in a Filipino Male: a Case Report and Review of the Literature



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ABSTRACT

Dupuytren's disease (DD) is a heritable, benign, chronic fibroproliferative process which affects the connective tissue of the palmar fascia. DD is rare among Asians with a prevalence of 0.004 to 0.032 percent. There are only 74 cases of DD among Asians identified in literature, and there are no published cases from the Philippines. We discuss a rare case of DD in a 60-year-old male Filipino presenting with bilateral loss of range of motion of the middle, ring, and little finger of both hands.

Key words: Dupuytren's contracture, Dupuytren's disease, Asia, Filipino, Case Report

INTRODUCTION

Dupuytren's disease (DD) is a heritable, benign, chronic fibroproliferative process which affects the connective tissue of the palmar fascia and can manifest itself as a clinically challenging disorder for both the patient as well as the surgeon. Dupuytren's contracture is an end consequence of the disease process.[1] The etiology of DD is still unknown. Researchers have hypothesized that a genetic problem is related to DD because of its heredity and racial predominance.[2] DD is common among Caucasians with a prevalence of 5.1 to 46 percent.

The disease is rare in Asians with a prevalence of 0.004 to 0.032 percent.[3]

The incidence and course of DD among Asians are mostly unknown as there are only a few reported cases in literature. A diagnosis of Dupuytren's contracture in a Filipino male seen in our clinic prompted a review of literature. To the best of our knowledge, this is the first reported case of DD occurring in a Filipino.

We discuss a rare case of DD in a 60-year-old male Filipino presenting with bilateral loss of range of motion of the middle, ring, and little finger of both hands.

Case Presentation

A 60-year-old male came to our clinic presenting with a loss of range of motion of the middle, ring, and little finger of both hands. Initially, he noticed tightening of the skin over the palmar aspect of both hands. Subsequently, he observed a painless, palpable mass in his palms. This was accompanied by difficulty in extending the little, ring, and middle fingers of both hands. The patient sustained a crushing injury to his left little finger after being caught in a car door years prior to the appearance of the above-mentioned symptoms. He is a non-diabetic with neither a history of epilepsy nor a family history of Dupuytren's contracture. We identified several risk factors associated with Dupuytren's disease. Our patient is a heavy laborer, smoker (20 pack years), and has an alcohol consumption of 600 g per week for 10 years. Family history is negative for Dupuytren's disease.

On physical examination, both hands revealed a positive Hueston tabletop test (Figure 1).

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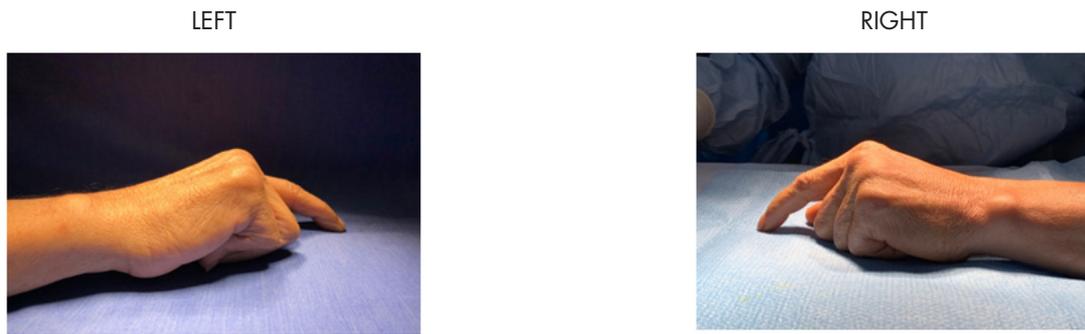


Figure 1. Case of Dupuytren's Contracture: Positive Hueston tabletop test on both hands. The patient is unable to lay the palm flat on a tabletop.



Figure 2. Case of Dupuytren's Contracture: Positive Hueston tabletop test on both hands. The patient is unable to lay the palm flat on a tabletop. of the left hand. **A.** Presence of MCP flexion contracture with PIP joint contracture of the middle, ring, and little fingers. A visible cord is seen over the palm. **B.** Boutonniere deformity of the little finger.



Figure 3. Case of Dupuytren's Contracture: Photograph of the right hand. Presence of MCP flexion contracture with PIP joint contracture of the middle, ring, and little fingers.

On the left hand, metacarpophalangeal (MCP) flexion contractures measuring 50 degrees, 30 degrees and 90 degrees on the middle, ring, and little fingers, respectively were observed. Proximal interphalangeal (PIP) joint contractures measuring 50 degrees on the middle and ring fingers and 135 degrees on the little finger were likewise seen (Figure 2a). There was a palpable central cord with retraction of the fingers. A boutonniere deformity of the little finger was also present from the previous crushing injury (Figure 2b).

The right hand also displays similar findings except for the presence of boutonniere deformity (Figure 3). The preoperative DASH scores were 32 and 31 for the left and right hands, respectively.

On further examination, there were no knuckle pads, Peyronie's disease or Ledderhosen's disease noted.

Surgery was performed on both hands under general anesthesia on two separate occasions. A Brunner-type incision was done to expose the palmar fascia. Intraoperative findings revealed central cords

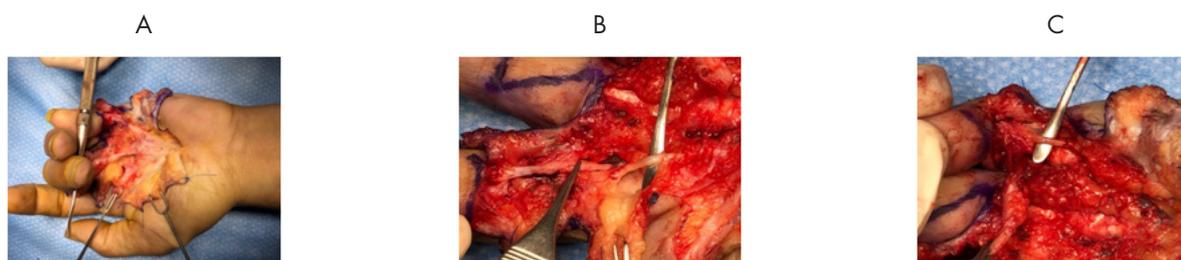


Figure 4. Case of Dupuytren's Contracture: Intraoperative findings (left hand). **A.** Central cords to the index, ring, and little fingers **B.** Spiral cord to the index finger. **C.** Lateral cord to the little finger



Figure 5. Case of Dupuytren's Contracture: Intraoperative findings (right hand). **A.** Central cords to the index, ring and little fingers **B.** Spiral cord to the index finger. **C.** Lateral cord to the little finger



Figure 6. Case of Dupuytren's Contracture: After regional fasciectomy of diseased cords (right hand). **A.** Full extension of the MCP joint of the little, ring, middle, and index fingers and PIP joint of the ring, middle, and index fingers. **B.** Boutonniere deformity limiting PIP extension of the little finger.

to the index, ring, and little fingers, a spiral cord to the index and a lateral cord to the little finger on the left hand (Figure 4). On the right hand, a central cord was noted to the ring finger and little finger (Figure 5).

Regional fasciectomy of the diseased cords was done on both hands, followed by gentle passive manipulation of the PIP and MCP joints. There was full extension of the MCP joint of the little, ring, middle, and index fingers and PIP joint of the ring, middle, and index fingers on both hands; however, the PIP joint of the little finger on the left hand could not be fully extended because of the boutonniere deformity (Figure 6, 7).

A primary wound closure was attempted, but a full-thickness skin graft from the thenar area was required to cover the soft tissue defect over the MCP crease of the little finger (Figure 8). After inset of the graft, the wound was dressed. A resting volar splint was placed to provide protection to the fresh wound. There were no immediate complications postoperatively. Two weeks postoperatively, sutures were removed and physical therapy was initiated.

Histopathologic examination showed multiple, light brown, irregular, rubbery tissues from 2.0 x 0.1 x 0.1 cm up to 3.5 x 0.2 cm. The microsections revealed benign fibrocollagenous tissue composed of

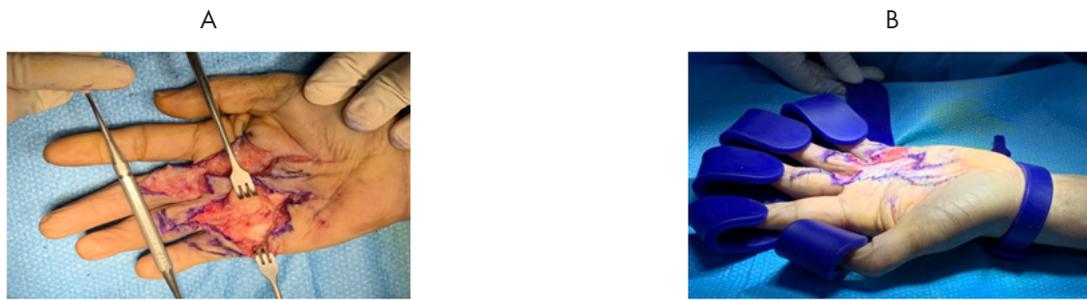


Figure 7. Case of Dupuytren's Contracture: After regional fasciectomy of diseased cords (left hand). **A.** Full extension of the MCP joint of the little, ring, middle, and index fingers and PIP joint of the ring, middle, and index fingers.

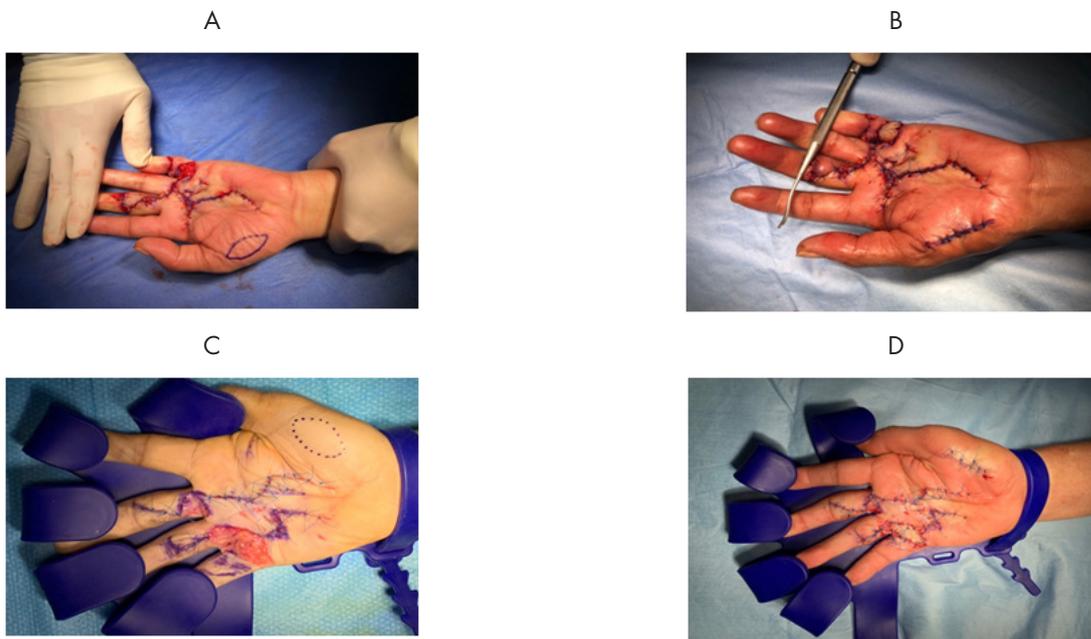


Figure 8. Case of Dupuytren's Contracture: Wound closure. **A.** Soft tissue defect over the MCP crease of the little finger (left). **B.** Full-thickness skin graft from the thenar area to cover defect (left). **C.** Soft tissue defect over the MCP crease of the little finger (right). **D.** Full-thickness skin graft from the thenar area to cover defect (right).



Figure 9. Case of Dupuytren's Contracture: Gross/microscopic findings. **A.** Multiple, light brown, irregular, rubbery tissues from 2.0 x 0.1 x 0.1 cm up to 3.5 x 0.2 cm. **B.** Bland fibroblasts on 40X magnification.

bland fibroblasts consistent with palmar fibromatosis (Figure 9).

Upon follow-up on the 3rd month post surgery, examination showed neither recurrence nor

extension of the disease. There were no wound complications. The patient is now able to fully flex and extend both his fingers except for the little finger on the left (Figure 10). DASH score of the left hand

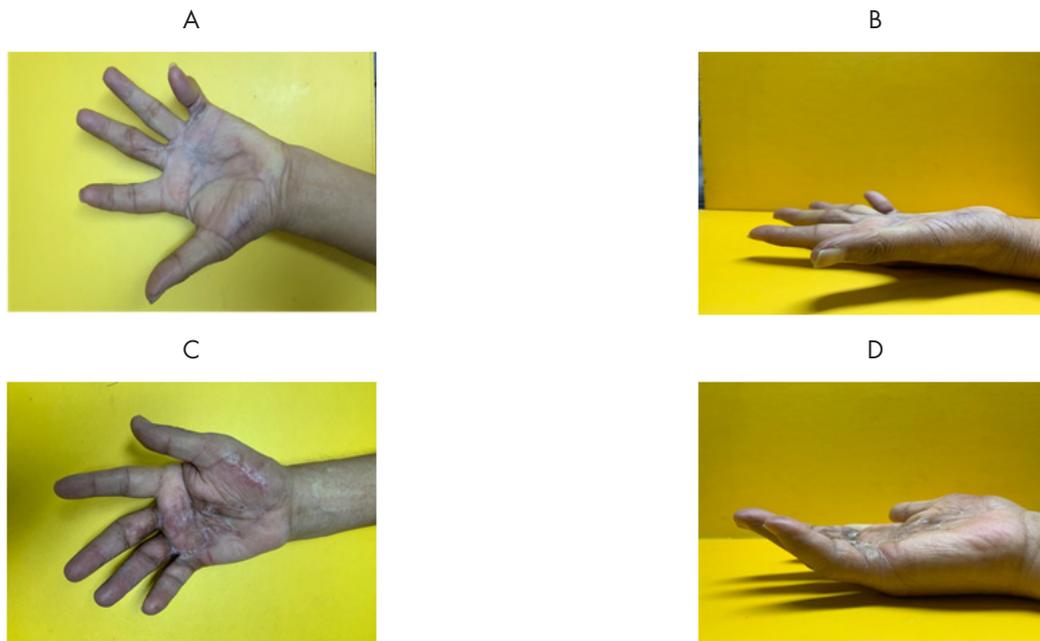


Figure 10. Case of Dupuytren's Contracture: Follow-up. **A, B.** Able to fully extend the index, middle, and ring fingers (left). **C, D.** Able to fully extend the index, middle, and ring fingers (right).

and right hand decreased from 32 to 21 and 33 to 22, respectively.

DISCUSSION

The diagnosis of DD in our patient was made due to the presence of bilateral flexion contracture over the middle, ring, and little fingers of both hands. Rayan and Moore classified the disease as typical DD and atypical DD. Typical DD is defined as a male Caucasian from Northern Europe exhibiting bilateral involvement and presence of ectopic lesions. The primary predisposing factor for this type is genetics. On the other hand, the atypical type has a random ethnic and gender distribution, unilateral involvement with no ectopic lesions. The predisposing factors linked to this type is history of surgery or trauma.[4] The salient features of alcoholism, heavy smoking, heavy labor, with no family history of DD in a Filipino suggests a diagnosis of an atypical DD.

Epidemiological data suggests that the highest prevalence of DD occurs in white northern Europeans with a reported prevalence of 32 to 46 percent. On the contrary, DD has an estimated prevalence of 0.004 to 0.032 percent among Asians which is 100–1,000 times lower than in western countries. [3] There are only 74 cases of DD identified in

literature, and there are no published cases from the Philippines (Pubmed, Cochrane, EMBASE).[4-8]

There is limited data in literature regarding the management of DD in the Asian population. A case series by Srivastava, et al. included 10 Indians operated at a hospital in England. He reported a success rate of 70% from radical or limited fasciectomy.[10] The largest series of surgical management was conducted by Abe, et al. which included 73 hands of 57 Japanese patients with DD. He observed recurrence of the disease in eight subjects (14%) and extension in nine (16%). Both recurrence and extension has been linked with ectopic lesions or involvement of the radial side of the hand.[12-14] Our patient was managed with radical fasciectomy with no complications and recurrence on follow up.

Slattery published a comprehensive review about DD among Asians. He mentioned that DD is a condition that is under-reported in the Asian population and it does follow a pattern that is yet to be ascertained. It appears that aside from familial predisposition and racial predilection, there may be other underlying factors for the development of disease in this population. Individual genetic susceptibility and risk factors such as diabetes, epilepsy, alcoholism, hand trauma, and heavy labor may have a role in the expression of DD in this population.[15]

CONCLUSION

We presented a rare case of atypical Dupuytren's in a Filipino male presenting with bilateral loss of range of motion of the middle, ring, and little finger of both hands managed with regional fasciectomy. Individual genetic susceptibility and risk factors such as diabetes, epilepsy, alcoholism, hand trauma, and heavy labor may have a role in the expression of DD in this population.

Ethical Considerations

The information that we have collected which may lead to the identification of the patient such as the name, date of birth, medical record number, geographic location, and phone numbers were not disclosed to comply with the principles of anonymity and confidentiality. Both written and verbal consent were given by the patient. The patient voluntarily agreed to provide information for this case which included gross photographs which were used for the purpose of research.

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